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A rare cause of testicular torsion: Lymphocytic infiltration in an adolescent with recently diagnosed B-ALL



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ABSTRACT

Testicular torsion is a commonly encountered medical emergency in children. A 10-year-old boy with diagnostically confirmed leukemia presented with new onset testis swelling. Scrotal ultrasound showed absent blood flow on the left, consistent with acute testicular torsion. The patient underwent left orchiectomy due to the testis being unsalvageable. Later pathology confirmed lymphoblastic infiltrates. A malignancy of the testicles is rarely associated with torsion and, in the setting of leukemia, suggests widespread disease. Due to the risk of scrotal violation, an inguinal approach is preferable for surgical exploration of the testicles in patients with a history of leukemia.

1. Background

In the United States, testicular torsion is a medical emergency in males, accounting for the most common cause of testicular loss. Testicular torsion is most often seen in young males under 25 years old with the incidence dropping dramatically past early adulthood. Testicular torsion typically presents on physical exam with acute onset severe testicular pain, often accompanied by nausea and vomiting. Here we describe a rare case of testicular torsion associated with lymphocytic infiltration of the testis in a patient recently diagnosed with B-cell acute lymphoblastic leukemia (B-ALL).

2. Case presentation

A Hispanic 10-year-old boy with no significant past medical history initially presented with 2 weeks of headaches, lethargy, nausea, and vomiting to an outside hospital with an elevated white blood cell count. Bone marrow cytometry later confirmed leukemia with an abnormal immature B-cell population. On hospital day 6, patient presented with overnight testicular pain and swelling on the left. Scrotal ultrasound was performed and showed absent flow to the left testis, consistent with acute testicular torsion (Fig. 1). Ultrasound was not suggestive of any other testicular abnormalities. The patient underwent emergent testicular detorsion but the left testis was not salvageable. A left orchiectomy followed by a right orchiopexy was performed. On gross examination, the left testis appeared hemorrhagic and edematous although no masses were identified. Pathologic examination of the testicles showed lymphoblastic infiltrates with marked vascular congestion and extensive involvement of the left testis, epididymis, and vas deferens (Fig. 2). Early



Fig. 1. Doppler ultrasound demonstrating testicular torsion of the left testis.

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Fig. 2. The left testis shows an interstitial hemorrhagic infiltrate with leukemic blasts (Figure A, $4 \times$). The lymphoid blasts surround the seminiferous tubules and show small cells with high nuclear-cytoplasmic ratio, irregular nuclear contours, and small nucleoli (Figure B, $40 \times$).

acute ischemic changes were noted in the seminiferous tubules with sloughing of cells. Right testis biopsy was subsequently performed and showed leukemic involvement.

3. Discussion

Testicular torsion is a common urologic emergency seen in the pediatric population. Testicular torsion has been known to occur in the setting of testicular malignancy. Published data, however, remains scant on the association between these two entities. While the incidence of testicular malignancy in the setting of torsion is rare, in one retrospective review by Uguz et al. that included 32 patients who had been diagnosed with testicular torsion, 2 of the patients (6.4%) were diagnosed with testicular malignancy – both germ cell tumors.¹

A small minority of boys with B-ALL will develop testicular disease either at presentation or during the disease course. Testicular involvement in the setting of B-ALL suggests widespread disease and is associated with a poor prognosis. Associated presenting features which may also predict the likelihood of testicular involvement are lymphadenopathy, the degree of anemia, thrombocytopenia, and splenomegaly. Most boys with testicular infiltrates tend to be asymptomatic. In a study by Nguyen et al. that included 20 patients with testicular leukemia, physical exam changes were noted in only 20% of the boys.² In our patient, there were no obvious preceding clinical or imaging findings to suggest testicular involvement.

With testicular malignancies, scrotal violation is a notable concern. In a systematic review of 1182 orchiectomy cases in patients with germ cell tumors from Capelouto et al. scrotal violation was not associated with significantly worse survival outcomes; however, local recurrence rates were found to be significantly higher in patients with stage I disease who had scrotal violation (2.9% vs 0.4%).³ Studies have looked at the role of fine needle aspiration cytology and testicular biopsy to evaluate for testicular involvement and guide management of the disease.⁴ Despite no studies reporting on local recurrence and survival

outcomes of testicular ALL, due to the higher concern of metastasis to the testicles in patients with a history of leukemia, an inguinal incision is preferred over a scrotal approach.

Education is imperative to understand the signs and symptoms of acute scrotal pain and should be considered an important conversation to families and patients with newly diagnosed and possible recurrent leukemia due to its predilection to the testicles. Although representing a common problem for pediatric urologists, a study that included 538 parents of mostly urology patients found that only about a third were familiar with testicular torsion.⁵

4. Conclusion

It is unclear whether pediatric patients with testicular malignancy are at increased risk for testicular torsion. The finding of testicular torsion in association with testicular infiltrates in patients with lymphoma or leukemia is rare. In patients with a history of leukemia undergoing surgical exploration of the testis, an inguinal incision is preferred to minimize the risk of potential scrotal violation.

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